

■ SHORT COMMUNICATION ■

IMPENDING SEPSIS DUE TO A RUPTURED PYOMYOMA WITH PURULENT PERITONITIS: A CASE REPORT AND LITERATURE REVIEW

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SUMMARY

Objective: Pyomyoma is an uncommon complication of benign leiomyoma. Here, we report a rare case of a perforated pyomyoma with purulent peritonitis.

Case Report: A 53-year-old postmenopausal woman presented with impending septic shock on arrival at our emergency department. Physical examination and imaging studies revealed a pelvic mass with peritonitis. Emergency exploratory laparotomy due to suspicion of a ruptured pyomyoma or malignancy revealed a spontaneously perforated large pyomyoma leaking copious purulent material directly into the peritoneal cavity. Total hysterectomy and bilateral salpingo-oophorectomy were performed. Histopathology revealed no malignancy. The patient recovered smoothly after combined antibiotic therapy.

Conclusion: Gynecologists should be aware of this rare emergency condition, especially when handling a patient with a history of leiomyoma uteri and if signs of infection or even sepsis are present. [*Taiwanese J Obstet Gynecol* 2005;44(1):75-79]

Key Words: leiomyoma, pyomyoma, sepsis

Introduction

Pyomyomas, also called infectious or suppurative leiomyomas, are a rare but serious complication of benign leiomyomas. This life-threatening event has a 21% mortality rate [1,2], even after aggressive treatment. The high mortality rate is probably due to lack of early recognition and appropriate management of the disease [2]. Pyomyomas have been reported during pregnancy, in the period after an abortion or birth, with instrumentation of a myomatous uterus (e.g. intrauterine device insertion and dilatation and curettage), with ascending uterine infection, with cervical stenosis, and

in menopausal women [2,3]. We demonstrate the crucial role of early recognition of this dreadful infectious disease, for which only concurrent surgical removal of the infected source and antibiotic therapy are life-saving interventions.

Case Report

A 53-year-old postmenopausal woman (gravida 2, para 1, spontaneous abortus 1) came to our emergency department because of sudden onset of lower abdominal pain, nausea, vomiting, constipation, and low-grade fever for 3 days. On arrival at our emergency department, her body temperature was 35°C, pulse rate was 135/min, respiration rate was 16/min, and blood pressure was 114/60 mmHg. She had a history of a symptomless uterine leiomyoma for more than 30 years to which she had paid no attention. In addition, she had intermittent low abdominal pain, fever, and weight loss of 3-4 kg of unknown cause during the past 6 months. She denied

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any sexual activity for more than 1 year or any history of uterine instrumentation. She had no history of hypertension, diabetes mellitus, or other medical illness.

Physical examination revealed abdominal distension and tenderness over the entire abdomen with rebound tenderness and muscle guarding. Pelvic examination showed an enlarged smooth cervix with mucoid discharge and a huge firm immobile mass in the posterior wall of the uterus. Transvaginal ultrasonography revealed an enlarged uterus of about $15.9 \times 10.9 \times 14.5$ cm with a large heterogeneous echogenic tumor located on the fundus of the uterus (Figure 1). Irregular anechoic areas were interlaced with calcified foci within the tumor, suggesting a cystically degenerated huge myoma with calcification. In addition, massive ascites were found on sonographic examination.

Computed tomography (CT) revealed an enlarged uterus with multiple nodules in the myometrium. The

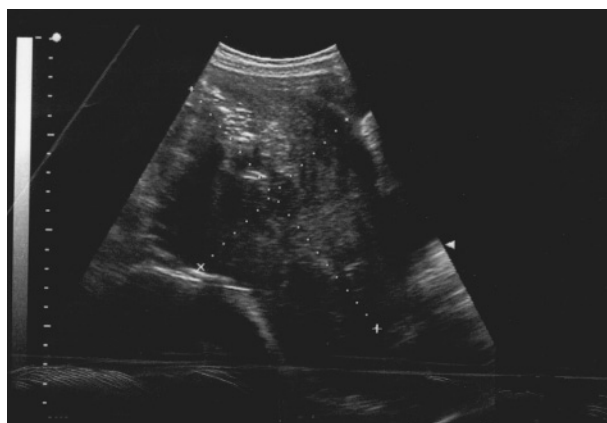


Figure 1. Transvaginal ultrasound showing an enlarged uterus with multiple irregular anechoic areas and calcified foci in the fundal tumor.



Figure 2. Computed tomography scan revealing intraperitoneal fluid, the presence of necrotic debris, and calcified deposits in a mass.

largest one presented with an ill-defined margin, predominantly with a necrotic component mixed with calcifications and solid foci (Figure 2). Ascites, dirty omental fat, and mild enhancement of the peritoneal membrane were also found. Abdominal tap revealed turbid ascites with a white cell count of $3.15 \times 10^6/\mu\text{L}$ with neutrophil predominance (60%), and Gram stain revealed a Gram-positive coccus. However, cultures yielded no bacteria. Routine hematologic and biochemical analyses of the blood showed 8.6 g/dL hemoglobin, a leukocyte count of 52,600/ μL with 81% neutrophils and 7.5% band forms, a platelet count of $7.48 \times 10^5/\mu\text{L}$, 140 mg/dL glucose, 81 mg/dL blood urea nitrogen, 2.5 mg/dL creatinine, and 42.4 mg/dL C-reactive protein. Urinalysis was unremarkable. Clinical and laboratory findings suggested impending sepsis with peritonitis of unknown origin R/O ruptured pyomyoma or malignancy and acute renal failure.

Exploratory laparotomy was performed shortly after admission due to persistent hypotension (systolic 90–100 mmHg) despite frequent fluid challenge, as well as to survey for malignancy. A large amount of purulent material was encountered (about 2,000 mL) initially and an abnormally enlarged, perforated left cornual leiomyoma was identified (Figure 3). Total hysterectomy and bilateral salpingo-oophorectomy were performed, and the peritoneal cavity was washed thoroughly with warm normal saline. Grossly, the uterus weighed 1,020 g, measured $19 \times 16 \times 12$ cm, and showed multiple leiomyomas with an extraordinary large left cornual leiomyoma. The left cornual leiomyoma measured about $12 \times 12 \times 10$ cm, was round, and contained massive necrotic tissue and multiple ossified tissue with perforation of the uterine serosa. In addition, it was completely separated from the endometrial cavity.

Histologic examination confirmed a pyomyoma without malignant transformation (Figure 4). Bacteriologic culture after 5 days disclosed that *Proteus mirabilis* was the offending organism. However, none of the blood cultures that had been collected grew any bacteria. Cefmetazole was administered for 5 days, which was switched to ceftizoxime and metronidazole according to sensitivity tests. The patient recovered quickly after the operation and was discharged in good condition on the 10th postoperative day.

Discussion

Pyomyomas are an unusual entity of uterine leiomyomas, but they invariably remain a life-threatening disorder and require surgical extirpation [4], even with the advanced surgical techniques and intensive intravenous



Figure 3. Enlarged uterus with multiple leiomyomas and a perforated leiomyoma on the left fundal side of the uterus.

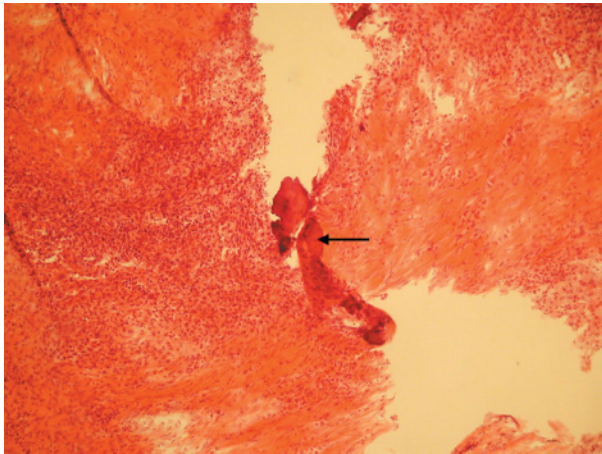


Figure 4. Section from the leiomyoma showing bundles of smooth muscle cells and infiltration of the tumor by numerous acute and chronic inflammatory cells; calcified material can also be seen (arrow).

antibiotic therapy currently available. It is postulated that pyomyomas are due to bacterial colonization of infarcted leiomyomas [5]. The offending organisms include *Clostridium* spp., *Staphylococcus aureus*, *Streptococcus milleri*, *Streptococcus hemolyticus*, *Proteus* spp., *Serratia marcescens*, *Actinomyces meyeri*, *Enterococcus faecalis*, *Edwardsiella tarda*, *Klebsiella pneumoniae*, *Candida parapsilosis*, *Peptostreptococcus tetradius*, and *Streptococcus agalactiae*. They may spread via direct invasion from an infected uterine cavity, neighboring structures, or lymphovascular routes [6].

Women who are highly vulnerable to such myomatous infections include pregnant women, postpartum or postabortion women, those with a history of instrumentation of the uterus, postmenopausal women, and immunocompromised women (e.g. with diabetes mellitus) [2,3], probably because the hemorrhagic or ischemic (infarcted) myomatous changes that

commonly occur during these periods put these women at greater risk of bacterial colonization [3]. Any infarcted leiomyoma can potentially be infected by even an occult infection [5]. Only this can explain why our patient, who had no history of diabetes mellitus, hypertension, prior uterine instrumentation, or recent childbirth, had an infected leiomyoma.

Since 1945, only 19 cases of pyomyoma have been reported (including the current report) (Table) [2,7–16]. Nine were related to pregnancy or abortion, seven occurred in postmenopausal women, and three were neither pregnant nor menopausal women. The modes of presentation and complications of pyomyoma vary. The most common presentations in the 19 cases reviewed were fever (84%), abdominal pain (57%), abdominal mass, and weight loss. Common complications included peritonitis (7/19), sepsis (5/19), and endocarditis (2/19). However, the interval between the initial onset of symptoms and diagnosis varied greatly. Pyomyoma can present with abrupt onset or may extend to a year of incubation. Patients are sometimes not aware of the condition. It can remain silent or nonspecific as an unidentified origin of bacteremia [3], as in our patient who had experienced unexplained low-grade fever and tolerable low abdominal pain. However, if a pyomyoma ruptures, the patient will experience severe peritonitis, which may present as an acute abdomen [4, 13]. Due to the low incidence, a rapid clinical diagnosis is usually difficult and the possibility of malignancy should never be excluded. In addition, prompt surgical intervention is required to correct the ongoing adverse complications that include pyoperitonitis, septicemia, and adult respiratory distress, and to prevent death [3, 16], since the mortality rate is high (15.8%, 3/19).

The differential diagnoses for a pelvic mass complicated with septicemia include tubo-ovarian abscess, pyometra with obstruction of the endocervical canal, gynecologic malignancy with invasion of the bowel subsequent to a perforated intestine and peritonitis, and pyomyoma [3]. The sonographic picture of a pyomyoma consists of an enlarged heterogeneously echogenic pelvic mass with solid and cystic components [10]. Characteristic CT findings of a ruptured pyomyoma with peritonitis include air and debris in the leiomyoma, discontinuity of the myoma wall, and intraperitoneal free air and ascites [7]. In addition, reformatted multiplanar sagittal and coronal images with contrast-enhanced CT may depict a more delicate picture and allow more accurate diagnosis of a pyomyoma with or without rupture [7]. The imaging studies in our patient showed these characteristics (Figure 2). The sonographic findings included a huge cystic degenerated leiomyoma with heterogeneous

Table. Summary of pyomyoma cases reported since 1945

Reference, year	Age (yr)	Underlying conditions	Presentation	Offending agent(s)	Treatment	Outcome	Complications
Cases related to pregnancy							
[2], 2002	33	Leiomyoma, cesarean delivery 6 d earlier	Fever, abdominal pain	<i>Candida parapsilosis</i>	ATH, antibiotic	Cured	Septic shock
[7], 2003	36	Leiomyoma, spontaneous abortion 1 wk earlier	Acute abdominal pain, fever	<i>Peptostreptococcus tetradus</i>	Myomectomy, use of antibiotic not mentioned	Cured	Peritonitis
[8], 2001	44	Leiomyoma, pregnancy	Fever	<i>Klebsiella pneumoniae</i>	Cefuroxime + metronidazole, cesarean section + myomectomy	Cured	Sepsis
[9], 1996	31	Pregnancy, intravenous drug abuser	Lower abdominal pain, constipation	<i>Staphylococcus aureus</i>	ATH + BSO, ampicillin + clindamycin + aztreonam	Cured	Peritonitis
[10], 1996	32	Elective abortion 10 wk earlier, leiomyoma	Lower abdominal pain, fever	<i>Enterococcus faecalis</i>	Cefoxitin, ATH + BSO, ampicillin + gentamycin + metronidazole	Cured	Peritonitis
[11], 1986	37	Leiomyoma and myomectomy 18 wk previously, spontaneous abortion 9 wk earlier	Fever, weight loss	<i>Streptococcus milleri</i>	Penicillin + streptomycin, ATH + BSO	Cured	Infective endocarditis
[12], 1957	29	3 wk after delivery	Fever, abdominal pain	Not mentioned	Penicillin + streptomycin + oxytetracycline, vaginal myomectomy	Cured	-
[13], 1963	34	Leiomyoma, pregnancy	Fever, abdominal pain	Gram-positive cocci	Penicillin + streptomycin, ATH + BSO	Cured	Preterm delivery, peritonitis
[14], 1986	29	Intrauterine device, pregnancy	Spontaneous abortion, amnionitis, fever	<i>Staphylococcus aureus</i> , <i>Serratia marcescens</i>	Ampicillin + gentamycin, ATH + BSO, nafcillin + clindamycin + cefotaxime	Cured	Ovarian thrombophlebitis, hemato-peritoneum
Premenopausal							
[3], 1990	49	Leiomyoma, habitual apicort juice injection	Fever, weight loss, lower abdominal pain	<i>Enterococcus</i> spp., <i>Staphylococcus aureus</i> , <i>Actinomyces meyeri</i>	Gentamycin + clindamycin + ampicillin	Died	Septic shock
[4], 1956	50	Cystoscopic removal of ureteral calculus 3 mo previously	Acute abdomen	Coliform group	ATH + BSO, tetracycline	Cured	Peritonitis
[15], 1999	46	Biliary stones, pancreatitis	Suprapubic pain, fever	<i>Edwardsiella tarda</i>	Amikacin switched to cefoxitin, ATH + BSO	Cured	Peritonitis
Postmenopausal							
[1], 2001	60	Leiomyoma, diabetes	Abdominal mass, weight loss, fever	<i>Streptococcus agalactiae</i>	Penicillin + amikacin, ATH + BSO	Cured	Endocarditis, DVT
[5], 1985	68	In good health	Pedal edema, abdominal mass, weight loss, mild fever	<i>Streptococcus</i> spp.	ATH + BSO, use of antibiotic not mentioned	Cured	Pedal edema
[6], 1976	59	Diabetes	Weight loss, fever	<i>Proteus</i> spp., <i>Clostridium</i> spp., <i>Enterobacteriaceae</i> , <i>Streptococcus</i> spp.	Penicillin + streptomycin, ATH + BSO	Cured	Abdominal wall fistulization
[16], 1999	75	Not mentioned	Fever, abdominal mass	<i>Streptococcus aureus</i>	ATH + BSO, antibiotic	Cured	-
[17], 1974	58	Leiomyoma, hypertension, diabetes	Shock, jaundice, fever	<i>Clostridium perfringens</i>	Penicillin	Died	Hemolysis, septic shock
[18], 1945	51	Uterine prolapse, hypertension	Sepsis, hypotension	<i>Streptococcus hemolyticus</i>	Sulfadiazine, subtotal hysterectomy + BSO	Died 8 hr postoperatively	-
Current case	53	Leiomyoma	Acute abdomen, fever, constipation	<i>Proteus mirabilis</i>	Cefmetazole, ATH + BSO, cefizoxime + metronidazole	Cured	Peritonitis, sepsis

ATH = abdominal total hysterectomy; BSO = bilateral salpingo-oophorectomy; DVT = deep vein thrombosis.

echogenicity as well as massive ascites. CT also revealed marked ascites and large leiomyoma uteri that contained focal necrosis and calcification, but there was neither intraperitoneal air nor air in the infected leiomyoma that may have directly led to a conclusive diagnosis of a ruptured pyomyoma.

In conclusion, the triad proposed by Greenspoon et al of a leiomyoma, bacteremia, and sepsis without a clear source of infection should always raise the suspicion of a pyomyoma [3]. Surgical removal of the infected myoma through either a hysterectomy or myomectomy accompanied by aggressive antibiotic therapy is the only life-saving intervention for this otherwise lethal condition (Table).

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